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Case report

Intramural tracheobronchial remnants associated with esophageal atresia: diagnostic aids

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Abstract. A patient with isolated esophageal atresia and congenital esophageal stenosis resulting from intramural tracheobronchial remnants (TBR) in the distal esophagus is presented. The esophagographic appearance and operative findings suggesting the presence of TBR are discussed.

Key words: Tracheobronchial remnants – Congenital esophageal stenosis – Esophageal atresia

Introduction

Congenital esophageal stenosis due to intramural tracheobronchial remnants (TBR) is an uncommonly encountered lesion [2, 4]. Since this rare anomaly may be associated with other congenital esophageal lesions, its diagnosis and treatment are important for a successful outcome. A case of esophageal stenosis due to intramural TBR associated with isolated esophageal atresia is reported to discuss the clinical features suggesting the presence of this rare occurrence.

Case report

A 1-day-old, 2,000-g female, after a normal delivery following a 37-week pregnancy, was referred to the Hacettepe Children's Hospital Department of Pediatric Surgery because of continuous regurgitation of saliva and choking during feedings. Passing a tube through the esophagus into the stomach was not possible. Radiologic examination revealed a blind upper esophageal pouch and a gasless abdomen. The patient initially underwent a gastrostomy. Subsequent evaluation with contrast material revealed a 1-cm-long gap between the atretic segments. Primary anastomosis of the esophagus was accomplished through a right-sided thoracotomy by an extrapleural approach. A 6 F feeding tube was easily passed through the esophagus into the stomach.

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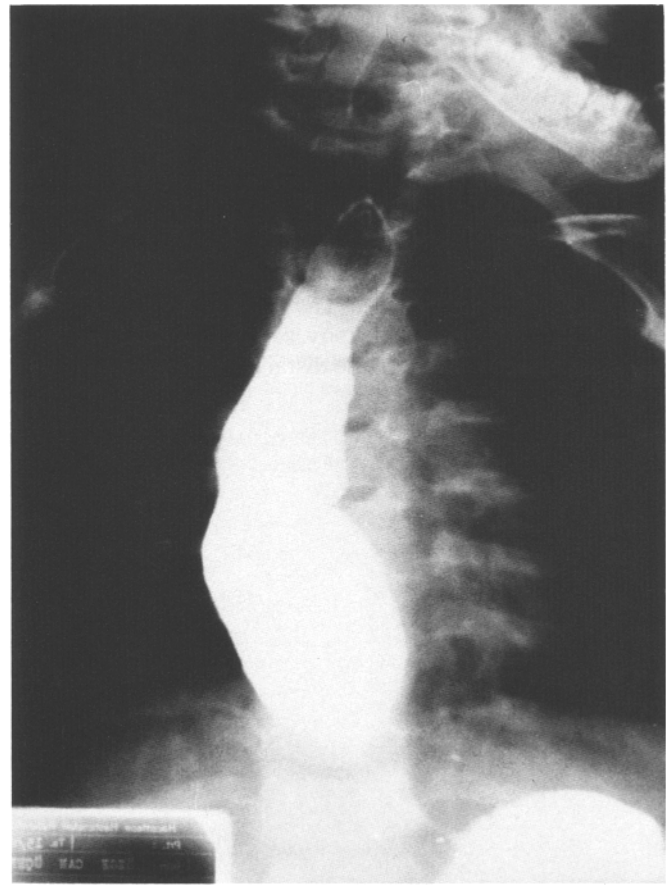

Fig. 1. Esophagogram showing tapered narrowing of distal portion of esophagus caused by TBR and dilatation proximal to the stenosis

At 10 months of age the patient was readmitted with inability to swallow. Esophagographic examination showed a tapered narrowing of the distal esophagus mimicking achalasia (Fig. 1), however, the rigid esophagoscope would not pass through the narrowed segment. Therefore, a laparotomy was performed with the initial diagnosis of esophageal stricture resulting from reflux esophagitis. During exploration, a size 7 Hegar dilator was passed through a gastrotomy into the distal esophagus. The dilator could not be passed through the distal esophagus 2 cm proxi-

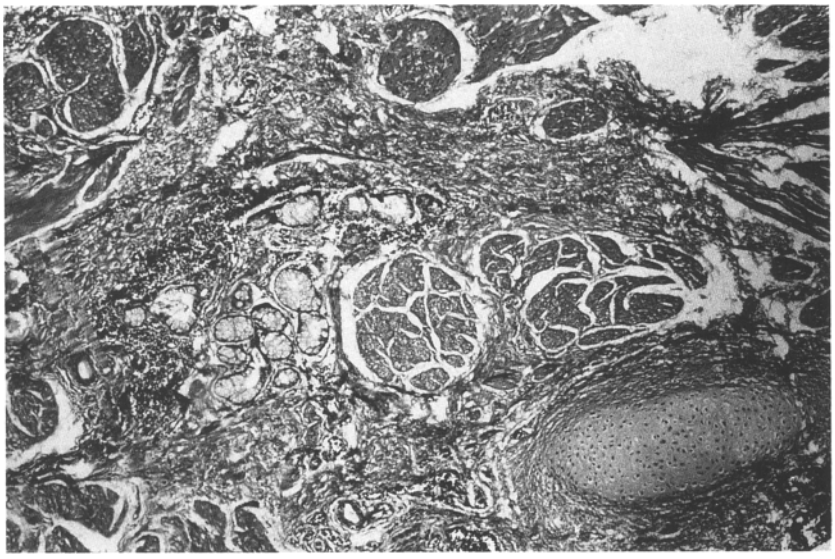

Fig. 2. Section from stenotic segment of esophagus showing islands of respiratory mucous glands and cartilaginous plates between esophageal musculature

mal to the esophagocardiac junction. A 0.5-cm narrowed esophageal segment was resected and esophageal anastomoses and a Thal fundoplication were performed. Histopathologic examination of the resected segment revealed TBR in the esophageal wall (Fig. 2). The postoperative course was uneventful.

Discussion

Congenital esophageal stenosis, which is accepted to be a variation of esophageal atresia, may present as a web, idiopathic muscular hypertrophy, or TBR [1, 3]. Intramural TBR are usually located in the distal third of the esophagus and presumably result from failure of the normal embryonic separation of the respiratory diverticulum from the foregut [4, 7]. Since they share a common embryologic basis, the association with congenital esophageal atresia and tracheoesophageal fistula (TEF) is not surprising. Several patients with proximal esophageal atresia and distal TEF and one with isolated esophageal atresia who additionally had TBR in the distal esophagus have previously been reported [2, 5, 7].

Unless the affected segment is resected, preoperative diagnosis of esophageal stenosis caused by TBR is very difficult. Since resection of the involved segment is required for treatment, some additional preoperative diagnostic procedures are necessary. The few previously reported cases of esophageal atresia and TBR revealed tapered narrowing of the distal esophagus during esophagographic examination that was suggestive of achalasia [2, 6]. Tapered narrowing should arouse the suspicion of the presence of TBR in patients who have esophageal atresia with or without TEF. Operative exploration of such patients should also include the passage of a dilator through the narrowed segment. If involvement of a short segment proximal to the cardioesophageal junction is detected, the possible presence of TBR in this segment should be considered.

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